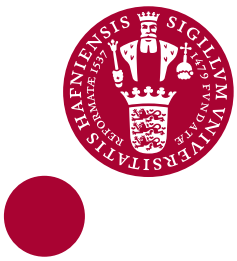

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BRIEF REPORT

Schistosomiasis Presenting as Recurring Sigmoid Volvulus in a Danish Man With an Inconspicuous Travel History—A Case Report

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A healthy 72-year-old Danish male presenting with recurring sigmoid volvulus was found to be infested with *Schistosoma mansoni*. No other explanation for recurring volvulus was found. A travel history 12 years ago, which included bathing in the Botswana Okavango delta for 10 minutes, revealed the likely time and place of infection. To our knowledge, this is the first reported case of recurrent sigmoid volvulus and chronic intestinal schistosomiasis in a patient from a nonendemic area.

Keywords. mansoni; schistosomiasis; travel; volvolus.

Helminthic infection caused by *Schistosoma mansoni* is a major cause of morbidity and mortality, with an estimated 54 million infected individuals in Sub-Saharan Africa [1]. Certain types of freshwater snails serve as intermediate hosts from which cercariae are released. Cercariae of *S. mansoni* swim up to humans, penetrate the skin, and migrate via the human liver and hepatic portal system to reside in the inferior mesenteric veins, where eggs are released through the intestine wall into the lumen. An immune reaction toward eggs called Katayama fever may present 2–8 weeks after cercarial exposure [2]. Over time, the immune response is downregulated, but a granulomatous reaction that results from egg deposition continues, leading to fibrogenesis and progressive organ damage. In chronic infection with *S. mansoni*, progression to hepatointestinal and hepatosplenic granulofibrotic disease normally takes 5–15 years to develop. It often presents with intermittent abdominal pain, loss of appetite, weight loss, diarrhea, and hematochezia. Rarely reported complications in patients with intestinal schistosomiasis include appendicitis [3], intussusception [4, 5], and intestinal strictures [6, 7].

Diagnosis of *S. mansoni* infection can be made by detection of eggs in stools, biopsies, or using polymerase chain reaction (PCR). Infection can be confirmed by serological test for schistosome antibodies in the blood, but this test cannot distinguish between active and previous infection. Praziquantel or Oxamniquine is used for treatment. A second dose may be given several weeks after the initial treatment, as developing schistosomules (the postpenetrational larval form of schistosomes) are less susceptible than adult worms. Monitoring for excretion of eggs is recommended up to 6 months after completed treatment, and retreatment may be required [2].

The symptoms in travelers with chronic hepatointestinal schistosomiasis may be diffuse or absent, and exposure to infected water several years earlier may not be mentioned during consultation. Therefore, chronic schistosomiasis in travelers often remains unreported or is made coincidentally during investigation for differential diagnoses like carcinomas, intestinal polyps, or portal hypertension [8,10]. We report a case of unexpected recurring sigmoid volvulus, most likely caused by intestinal schistosomiasis, in a previously healthy 72-year-old Danish man whose medical record, before this admission, only included hypertension. To our knowledge, this is the first reported case of recurrent sigmoid volvulus in a patient from a nonendemic area and the second case worldwide [11].

CASE PRESENTATION

A healthy 72-year-old male was referred to the department of abdominal surgery with 8 days of constipation and diffuse cramping abdominal pain, with postprandial worsening and abdominal bloating in the last 2 days. There was no nausea or vomiting. He had self-medicated with laxatives without effect. The patient used Amlodipine (a calcium channel blocker) for hypertension, which in some elderly patients can cause constipation, but had tolerated this for many years with no prior history of constipation. The patient had no other chronic conditions or neurologic disease and didn't take any other medications.

Examination revealed an extended tympanic abdomen, palpation caused only mild pain, and auscultation revealed high-pitched and metallic sounds. Blood tests, including liver panel, C-reactive protein, and eosinophil count, were normal, except for low potassium. An abdominal computed tomography (CT) scan with contrast showed sigmoid volvulus, with a dilated

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sigmoid measuring 17–18 cm in diameter; the liver was unremarkable, and no other pathology was found.

An acute colonoscopy confirmed massive dilation of the sigmoid colon, and the mucosa appeared normal endoscopically. Substantial fecal masses made further passage impossible 50 cm from the anus. Colonoscopic decompression was performed with good results.

Later attempts of full colonoscopies were initiated 3 times but had to be disrupted due to incomplete emptying of the colon 50 cm from the anus. A flat sessile polyp in a villous area 50 cm from the anus was identified. It was not accessible for normal polypectomy, and endoscopic mucosal resection (EMR) was planned at a later stage. A CT-colonography was then performed to examine the right colon, but this also proved insufficient and did not show any additional pathology.

The patient was re-admitted 3 months after the initial presentation with 3 days of constipation and abdominal bloating. A CT scan verified recurring sigmoid volvulus with a "whirl sign" (mesenteric vessels creates the whirls where the bowel rotates around its mesentery) just before the cecum, which was dilated to 17 cm in diameter. Colonoscopic decompression was performed.

Taking the relapsing volvulus, the substantial risk of recurrence, and the need to examine the sessile polyp into account, together with the fact that the patient continued to travel and stay in uninhabited areas where medical and surgical emergency care where unavailable, it was decided to do a laparoscopic sigmoid colectomy.

Surgical Findings

A long and dilated sigmoid, including a long mesentery and effects of recurrent volvulus (fibrotic thickening of the peritoneum at the mesentery), was found. The polyp was not visible during surgery, and no other pathology was found during endoscopy. A colon resection of around 50 cm, including the affected segment, was performed and sent to the pathology department for further examination.

Pathology Findings

Macroscopic examination showed a colon resection of 50 cm, including 19 cm with thinning of the mucosa, colonic diameter up to 14 cm, and a wall thickness down to 3 mm. This area showed multiple slightly elevated polyp-like elements with a diameter of up to 5 mm. Microscopic examination included an adenoma with low-grade dysplasia. Underlying the submucosa and tunica muscularis, non-necrotizing granulomas were found. In 1 of these, a centrally located egg with a lateral spine characteristic of *S. mansoni* was found (Figure 1). A diagnosis of chronic intestinal *S. mansoni* infestation was made, and no other cause of sigmoid volvulus was found.

Referred to the Department of Infectious Diseases, Copenhagen University Hospital, the patient revealed that he had been traveling for 3 weeks in Botswana in 2005. There he had canoed in the Okavango Delta and bathed for 10 minutes. He could not remember having any symptoms during or after this trip, and specifically no diarrhea, pruritus, or asthmatic symptoms.

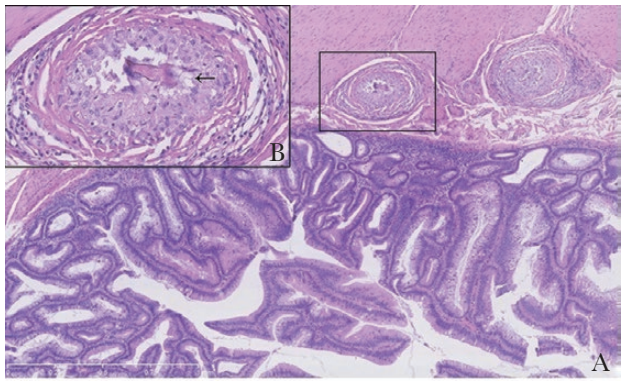

Figure 1. The microphotograph of an adenoma with low-grade neoplasia with non-necrotizing granulomas in the underlying submucosa, where in the center of one of these an egg (which is not yet calcified, arrow) with some parts of the embryo is seen. The egg is elongated and possesses a lateral spine characteristic of *S. mansoni* (H&E) 5x (A) and 40x (B).

Repeated blood tests showed normal liver function tests, CRP, and eosinophil count. Enzyme-linked immunosorbent assay (Novagnost Schistosoma mansoni, Siemens Diagnostics, Marburg, Germany) for schistosoma antibodies in serum was positive at 18.9 units of IgG (positive > 11.5 U; negative < 8.5 U). As expected, urine was without schistosoma eggs. Formalin-fixed tissue samples from the partial colectomy tested negative using a specific in-house PCR (Department of Mycology and Parasitology, Statens Serum Institut, Copenhagen, DK). Ultrasonography of the liver showed no hepatosplenic pathology.

Treatment with praziquantel (Biltricide), 2 doses of 20 mg/kg, was given and was tolerated well by the patient.

Six months after the sigmoid colectomy, a control colonoscopy showed no signs of intestinal pathology, but random biopsies presented granulomas with a centrally located egg-like structure compatible with *S. mansoni* eggs, but without visible lateral spine, representing empty remnants. Biltricide treatment was repeated.

No gastrointestinal symptoms were observed in the follow-up period, 7 months after the sigmoid colectomy.

DISCUSSION

To our knowledge, this is the first case of recurrent sigmoid volvulus in a patient from a nonendemic area incidentally found to have colonic schistosomiasis, and it is the second case worldwide [10]. In both cases, infection was not suspected before the sudden onset of volvulus. Our patient had not noticed any symptoms during his travels in Botswana 12 years ago, nor had he experienced symptoms in the following years. In both cases, diagnosis was made after parasitic eggs were found in the resected colon, and later serological testing verified infection

with *S. mansoni*. In the case from Mourra et al., a stool examination revealed schistosomal ova [9]; however, microscopy of stools was not performed in our case. We found a limited number of egg granulomas, and PCR on the tissue material was negative. As the PCR is highly sensitive, this suggests that eggs were empty shells devoid of miracidia, although a wide spacing of eggs in the mucosa could have contributed. Furthermore, neither CT scan nor ultrasonograms showed any sign of liver fibrosis. This indicates that light infections, which are most common in travelers, may potentially cause severe disease manifestations, as they are not recognized before major symptoms appear [10].

Whether the suspicion and serological confirmation of intestinal *S. mansoni* infestation at first visit would have led us to a more conservative approach using praziquantel and deflation as the primary strategy, without colon resection, is uncertain. In our case, the mesentery might already have been permanently long and loose, and the relapse of volvulus unavoidable without surgery. Unfortunately, no tissue samples with pathology directly affecting ganglia and/or nerve fibers were evaluated, and consequently, whether the peristalsis of the sigmoid colon was too damaged by inflammation was not addressed.

Our patient did not match the clinical picture of the regular volvulus patient, in which volvulus most often occurs, where the patient develops a large, elongated, relatively atonic colon, particularly in the sigmoid segment, because of long-standing chronic constipation. In our case, the patient was relatively young, in good health, with no substantial comorbidities. He had been an avid traveler and had visited multiple countries known to harbor schistosomiasis, but the diagnosis was initially missed because of the long timeline, the lack of prior symptoms, and the unimaginable possible association between schistosomiasis and volvulus. Still, the surgeon found him an "atypical" patient suffering from sigmoid volvulus.

Tourism is rising in many parts of the world, increasing exposure-related behaviors, including contact with infested fresh water. The risk of acquiring schistosomiasis is often unknown or neglected by travelers, either due to ignorance or to a perception of low risk of severe illness in light infections [11]. Our case underlines the importance of giving patients information about relevant travel exposures in advance of travel to endemic areas and exploring travel activities and exposures in patients presenting with symptoms of uncertain origin. In addition, serological testing after travel with exposure to fresh water in relevant geographical areas should be recommended as a screening method in order to identify and treat infection before surgery-demanding pathology develops.

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